

Psychological Adjustment of Children with Ambiguous Genitalia (Disorder of Sex Development) and Their Parents

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ABSTRACT

Background: Potential long-term sequel of ambiguous genitalia and its surgical correction does not only include functional problems for example incontinence or difficulties in voiding, but also cosmetic problems. Psychological adjustment and self-appraisal after these surgeries are sequel that have been insufficiently studied. These difficulties often evolve long after surgical repair as children grow to adulthood. **Subjects and methods:** A sample of 26 patients with ambiguous genitalia who had their first surgical intervention more than 5 years before were recruited. The children were assessed to find out the degree to which the appearance is upsetting, how it is related to negative evaluation of self, the degree to which it leads to avoidance; and their parents were assessed for the presence of grief and its stage. **Results:** The surgical condition was upsetting in 38.5% (n=10). It lead to negative evaluation of self in 23.1% n (and avoidance in 38.5% (n=10) 6); as for parents, It was found that 15.4 % (n=4) suffered denial; 53.8% (n=14) suffered depressive mood whereas 30.8% (n=8) suffered depressive behavior. Guilt was absent whereas anxiety was present in all parents. Risk factors for psychological maladjustment in both children and their parents included female sex, longer duration of illness, repeated surgeries, severe hypospadias, more residual problem or cosmetic defect. Presence of more depressive or anxiety symptoms in parents was positively correlated to upset and negative view of self in the child. **Conclusion:** It is thus concluded that outcome of surgery for ambiguous genitalia extends far beyond postoperative, functional and local cosmetic complications and that psychological outcome and patient satisfaction have long been overlooked and that proper follow up and management of those patients should include psychological care for the children and their parents over a long period of time that may extend into adolescence.

Key words: Ambiguous genitalia, Psychological adjustment, Disorder of sex development.

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INTRODUCTION

A newborn infant with ambiguous genitalia is a complex enough problem to unravel without any further clouding by confusing terms. The nomenclature 'intersex', 'hermaphrodite' and 'pseudohermaphrodite'

is anachronistic, unhelpful, and perceived to be pejorative by some affected families. In its place, a consensus statement recommends the term 'disorder of sex development' (DSD), a generic definition

encompassing any problem noted at birth where the genitalia are atypical in relation to the chromosomes or gonads. The karyotype is used as a prefix to define the category of DSD, replacing the arcane terminology of male or female pseudohermaphroditism (now known as XY DSD or XX DSD, respectively). The new nomenclature has spawned a simple and logical classification of the causes of DSD. It includes gonadal dysgenesis and novel defects in steroid biosynthesis¹. In other words, disorders of sex development are medical conditions in which the development of chromosomal, gonadal, or anatomic sex varies from normal and may be incongruent with each other².

In a study on an Egyptian sample, 208 patients with ambiguous genitalia were recruited from the genetic clinic from 2000 to 2005. Results showed that 46XY DSD was more common than 46XX DSD constituting 65.9% of total cases. Consanguinity was high with 61% in the affected families; however, only 21 cases had a positive family history. There was preference of male sex of rearing (regardless of karyotype), despite a severe degree of ambiguity³.

Hypospadias is one of the most common correctable birth anomalies and second only to cryptorchidism as the most common malformation of the male external genitalia. The incidence of hypospadias is approximately 1 of 250 to 1 of 300 live births. It occurs prenatally during formation of the external genitalia in boys⁴. This anomaly can be defined as an arrest in normal urethral development resulting in ectopic opening of the meatus. The position of the urethra determines the severity of the anomaly. Further, it is characterized by ventral curvature and open prepuce.

Milder forms such as distal and penile are the most common⁵. In a population-based study conducted in Canada between 1980 and 2007, the total number of male pregnancy and birth records was 130,796. The total number of cases of hypospadias was 995, yielding an incidence of 0.76%. The severity of hypospadias was graded as glanular in 428 (77.8%); coronal in 77 (14%); penile shaft in 34 (6.2%); and proximal to the penile shaft in 12 (2.2%). The severity of hypospadias was not graded in 445 cases⁷. The American Academy of Pediatrics considers the age between 6 and 12 months to be the best time for surgery for hypospadias⁷.

Female infants with classic congenital adrenal hyperplasia (CAH) typically have ambiguous genitalia at birth because of exposure to high concentrations of androgens in utero, and (CAH) is the most common cause of ambiguous genitalia in 46XX infants. Characteristic findings include an enlarged clitoris, partly fused and rugose labia majora, and a common urogenital sinus in place of a separate urethra and vagina. The internal female organs, the uterus, fallopian tubes, and the ovaries, are normal. Glucocorticoids treatment is recommended only for those with symptoms and aims to reduce hyperandrogenism.

Glucocorticoid treatment is indicated in children with androgen excess. It is given in doses sufficient to suppress adrenal secretion partly, without total suppression of HPA axis⁸. The surgical management of children born with ambiguous genitalia is complex. The Joint European Society for Pediatric Endocrinology and Lawson Wilkins Pediatric Endocrine Society recommend that the surgery should be done in virilised girls with classic CAH at age 2-6 months

because it is technically easier than at later ages⁸.

To perform corrective genital surgery for cases of intersex disorders, now designated as disorders of sex differentiation, it requires far insight into their problems that are faced even many years after the surgery. A number of surgical procedures need to be performed before the final outcome is attained⁹.

Over the years, outcome of corrective surgery is studied after a short period of follow up of 1 year duration according to the standard protocol¹⁰ and is mostly concerned with early surgical complications and short-term functionality¹¹⁻¹². Few studies were concerned with psychological outcome in these patients. A study by Schonbucher and his colleagues stated that psychosocial and psychosexual development of children and adolescents after hypospadias surgery has rarely been investigated and that while the results of the few studies available are altogether contradictory, they also indicate that hypospadias patients suffer from specific problems like negative perception of genital appearance. Therefore, they should be offered long-term follow-ups and psychosocial support until they reach young adulthood¹³. In Addition, psychosocial and clinical risk factors have hardly been examined¹³. On the other hand, it was found that psychological adjustment is not compromised in females with virilized genitalia who are treated early in life and reared as females¹⁴.

The researchers aimed in this study to investigate the psychological adjustment of children and their parents after surgical interventions and to find out about possible risk factors for maladjustment.

SUBJECTS AND METHODS

Twenty six patients with ambiguous genitalia in late childhood and adolescence, who were operated upon for the first time more than 5 years ago, were recruited from the outpatient clinic of pediatric surgery, Ain Shams University over a period of six months (April, 2008 till September, 2008).

They were interviewed using a structured questionnaire adapted from **Body Dysmorphic Disorder Examination Self Report**¹⁵. It is composed of 18 questions rated on a 6 point rating scale that assess the degree to which the appearance is upsetting (Q3-Q10), how it is related to negative evaluation of self (Q11-Q14), the degree to which it leads to avoidance (Q16-Q18) in situations where it could be noticed. A score on any item >4 is significant. Moreover, sum of all scores yields a total score of degree to which the appearance feature psychologically impacts the patient. Also sum of scores of questions specified before yields a score on upset by appearance feature, negative evaluation of self and avoidance respectively.

Their parents were also interviewed using a 20 items questionnaire designed by the researcher to assess the presence of grief and its stage if present based on the clinical picture of grief¹⁶. Symptom severity was rated on a 3 point rating scale. The scale yields a total grief severity score and scores for: Denial (Q1, 2, 3), Anguish: depressive symptoms (Q4-11); guilt (Q12, 13); anxiety (Q14-16); Consequent relation to the child (Q17-20).

Data Analysis

Qualitative variables were described in number and percentages and mean±

standard deviation (SD) if quantitative. Student T and ANOVA tests were used when comparing quantitative data between 2 or more than 2 groups respectively. Bivariate analysis was carried to find correlation between quantitative variables while chi square test was used to find out about associations between variables. P value was always set at 0.05. All data manipulation and statistical analyses were performed using SPSS (Statistical Package for Social Sciences) version 11.

RESULTS

The mean age of the 26 patients was 10.92 ± 1.76 st.d (range 8–14 years). Twenty of them were males (76.9%) and 6 were females (23.1%). All patients are in main stream education. Among the sample, 14 patients (53.8%) come from rural areas. Descriptive clinical data of the sample are listed in table (1):

All patients underwent their first corrective surgery at least 5 years ago, but they needed repeated surgical intervention mean 3.85 ± 1.74 st.d. (Range: 2-9). Late outcome of the surgery was assessed from both their parents' as well as their surgeon's view. None saw a worse or even no change outcome but complete cure in this sample was only found in 2 patients (7.7%). There was a very highly significant association between both views ($P=0.003$). Also the presence of residual functional problem and cosmetic defect was assessed. Mild cosmetic defect was found in 76.9% ($n=20$) of the sample while severe defect was found in 7.7% ($n=2$). Figures 1, 2, 3 & 4 show sample images for 2 patients' external genitalia before and after the surgery respectively. The only residual functional problem encountered in this sample was incontinence of urine in 30.8% ($n=8$).

Figure 1:



Figure 2:



Figure 3:



Figure 4:

Using Chi Square statistical test, there was a significant association between residual

problem and residual defect ($P=0.04$). Results are summarized in (table 2).

Psychological impact of the patients' surgical condition was assessed using the structured questionnaire adapted from "Body Dysmorphic Disorder Examination Self Report"¹⁵. Results showed that the surgical condition was upsetting in 38.5% ($n=10$). It lead to negative evaluation of self in 23.1% n (6) and avoidance in 38.5% ($n=10$) mean total score was 36 ± 18.4 std.; means and std. for subscales were (9.53 ± 6.6 ; 9.53 ± 5.01 ; 3.69 ± 3.58) respectively.

Table (1): Descriptive clinical data of the sample:

Age at diagnosis	mean: 0.38 mths	std.: ± 0.94	range: 0-3mths.
Age at 1 st . surgery	mean: 31.54mths	std.: ± 24.53	range: 2-72mths
Diagnosis	Mild to moderate hypospadias: n (2) f (7.7%)	Severe hypospadias n (18) f (69.2%)	Virilisation n (6) f (23.1%)
Hormonal state	Normal n (10) f (38.5%)	5 α reductase deficiency n (10) f (38.5%)	Congenital adrenal hypoplasia n (6) f (23.1%)

Table (2):

	Outcome parents' view		Outcome surgeon's view		
cured	n (2)	f (7.7%)	n (2)	f (7.7%)	X^2 : 26.00 P : 0.003*
improved	n (24)	f (92.3%)	n (24)	f (92.3%)	
	Residual functional problem		Residual cosmetic defect		
yes	n (8)	f (30.8%)	n (22)	f (84.6%)	X^2 : 6.28 P : 0.04*
No	n (18)	f (69.2%)	n (4)	f (15.4%)	

Psychological impact on parents was assessed using a questionnaire designed by the researcher to assess the presence of grief and its stage if present based on the clinical picture of grief¹⁶. Parents' mean total grief severity score was 23.85 ± 6.68 std. mean score on denial 4.38 ± 1.17 ,

depressive symptoms 9.15 ± 4.73 ; guilt 1.46 ± 0.86 and anxiety 5.00 ± 0.89 . It was found that 15.4 % ($n=4$) suffered denial; 53.8% ($n=14$) suffered depressive mood whereas 30.8% ($n=8$) suffered depressive behavior. Guilt was absent whereas anxiety was present in all patients. Regarding

consequent relation to the child parents self reported over protection in 53.8% (n=14); permissiveness in 30.8% (n=8); over criticism in 7.7% (n=2). All patients, however, reported sometimes avoiding the child so as not to be reminded of his condition.

Data were then analyzed to find any relation between sociodemographic or clinical parameters and psychological consequences on both children and their parents. There was a very highly significant difference in total grief & anxiety symptoms being higher among parents of female patients; similarly, there was a

highly significant difference in denial and depressive symptoms being higher in parents of female patients. Age of the patient during the study was significantly negatively correlated to the degree of guilt experienced by parents (table 3).

On the other hand, there was a highly significant difference in negative view of self between males and females being higher in females (p=0.04) while age of the patients during the study was highly and positively correlated to a degree to which appearance is upsetting and to avoidance (p=0.0001 for both). The details are shown in (table 4).

Table (3): Relation between sociodemographic variables and grief parameters in parents

		Total grief	Denial	Depressive symptoms	guilt	anxiety	Relation to child
Gender	male	21.7±6.09	4.1±1.17	7.8±4.49	1.3±0.92	4.7±0.8	4.8±0.75
	female	31.0±1.55	5.33±0.52	13.67±1.86	2±0.1	6±0.1	5±1.55
	t (df)	-3.65 (24)	-2.49(24)	-3.08(24)	-1.83(24)	-3.91(24)	-0.43(24)
	p value	0.001*	0.02*	0.005*	0.08	0.001*	0.6
Age	r	-0.23	-0.29	-0.13	-0.39	0.001	-0.28
	p value	0.2	0.14	0.5	0.04*	1	0.1

Table (4): Relation between sociodemographic variables and body dysmorphic parameters in children

		Total B Total BDD	Upsetting	Neg. view of self	avoidance
Gender	male	35.3±19.65	9.5±6.95	8.2±3.83	4.4±3.68
	female	38.33±14.8	9.67±6.09	14±6.2	1.33±2.07
	t (df)	-0.34(24)	-0.05(24)	-2.81(24)	1.93(24)
	p value	0.7	0.95	0.01*	0.06
Age	r	0.73	0.68	0.34	0.74
	p value	0.7	0.0001*	0.08	0.0001*

Clinical variables were also investigated for probable significant relation to psychological adjustment of patients and their caregivers. There was a significant difference between mild to moderate hypospadias patients, severe hypospadias

patients and virilization with respect to negative view of self and avoidance in patients; and with respect to all items of grief except guilt and relation to the child in caregivers. In all cases post Hoc tests showed significant difference existed with

mild to moderate as compared to severe or as compared to virilization. Details are shown in (tables 5 and 6)

Presence of residual functional problem and/or residual cosmetic defects and its degree lead to a very highly significant difference in all grief scores except guilt which was significant and relation to the

child which was insignificant. Again post Hoc test showed that significance existed between scores with no problem or defect as compared to presence of functional problem or severe defect. Same applies for scores on body dysmorphic self report. Details are shown in (tables 5 and 6)

Table (5): Relation between clinical variables and grief parameters in parents

		Total grief	Denial	Depressive symptoms	guilt	anxiety	Relation to child
Age at diagnosis	r p	0.4 0.04*	0.36 0.06	0.3 0.1	0.26 0.18	0.47 0.01*	0.15 0.4
Age at 1st. surgery	r P	-0.47 0.01*	-0.22 0.2	-0.48 0.01*	-0.12 0.5	-0.48 0.01*	-0.07 0.7
No. of surgeries	r P	-0.02 0.9	0.07 0.7	-0.04 0.8	-0.43 0.02*	0.2 0.3	0.17 0.39
Diagnosis	f(df) p	7.72 .003*	4.56 0.02*	5.96 0.008*	2.55 0.10	9.20 0.001*	0.13 0.87
Outcome parent's view	x ² p	2.40 0.13	3.32 0.08	2.75 0.11	0.84 0.36	2.91 0.10	0.05 0.82
Residual problem	f(df) p	-5.16 0.0001*	-4.17 0.0001*	-4.75 0.0001*	-2.3 0.03*	-3.39 0.002*	-0.53 0.5
Residual defect	f(df) p	3.71(2) 0.04*	7.2(2) 0.004*	3.78(2) 0.03*	0.42(2) 0.6	3.63(2) 0.04*	1.18(2) 0.3

Table (6): Relation between clinical variables and body dysmorphic parameters in children

		Total BDD	Upsetting	Neg. view of self	avoidance
Age at diagnosis	r p	-.08 .68	-.09 .63	.03 .84	-.43 .02
Age at 1st. surgery	r p	-.57 .002	-.41 .03	-.62 .001	-.27 .18
No. of surgeries	r p	.46 .01	.61 .001	-.008 .96	.44 .02
Diagnosis	f(df) p	1.98 .16	.74 .48	7.17 .004*	4.22 .02*
Outcome parent's view	x ² p	4.14 .05	1.54 .22	5.87 .02*	2.42 .13
Residual problem	t p	-1.94(24) 0.06	-1.85(24) 0.07	-3.1(24) 0.005*	0.64(24) 0.5
Residual defect	f(df) p	13.18(2) 0.0001	13.25(2) 0.0001	2.27(2) 0.1	4.71(2) 0.01

Scores on both grief variables and body dysmorphic self report variables were investigated for correlation between grief in

parents and upset or negative view of self and avoidance in children using pearson bivariate analysis. Negative view of self

was positively correlated to total grief of parents and results were of statistical significance ($P=0.01$). There was a positive correlation to depressive symptoms in parents and results were of very high

statistical significance ($P=0.002$). Upset about appearance of genitalia was positively correlated to anxiety of parents and results were of statistical significance ($P=0.02$). Results are shown in (table 7)

Table (7): Correlation between grief parameters and body dysmorphic parameters:

		Total BDD	Upsetting	Neg. view of self	avoidance
Total grief	r	0.30	0.24	0.47	-0.02
	p	0.13	0.22	0.01*	0.88
Denial	r	0.19	0.20	0.25	-0.06
	p	0.33	0.30	0.21	0.74
Depressive symptoms	r	0.35	0.27	0.57	-0.002
	p	0.07	0.17	0.002*	0.99
guilt	r	0.10	0.06	0.20	-0.004
	p	0.62	0.74	0.32	0.98
anxiety	r	0.42	0.40	0.35	0.22
	p	0.02*	0.04*	0.07	0.27
Relation to child	r	-0.37	-0.29	-0.34	-0.31
	p	0.05	0.13	0.08	0.11

DISCUSSION

Over a long period, much attention has been paid to the short-term results of corrective genitoplasty such as urethral strictures and fistulas. Therefore although thousands of articles on hypospadias were published, only a few were involved in the long term outcome of hypospadias let alone psychological outcome. When virilisation is considered even much less articles were published. The researchers aimed in this study to investigate the psychological adjustment of children with ambiguous genitalia and their parents after surgical interventions and to find out about possible risk factors for maladjustment.

Our sample was composed of 26 patients with ambiguous genitalia in late childhood and adolescence, which were operated upon for the first time more than 5 years ago. They were recruited from the outpatient clinic of pediatric surgery, Ain Shams University over a period of six months.

Their mean age at first surgery was 31.5 months (2 years and 7 months); age range 2-72 months. This is not comparable to most studies on corrective genital surgeries and to the current American Academy of Pediatrics recommendation to perform hypospadias repair at age 6 to 12 months¹⁷. Wide age range might reflect the need for raising the awareness of people in rural areas as well as improving health care provided to newborns for early detection and intervention in those areas.

Mean age at the time of the study was 10.92 ± 1.76 st.d (range 8–14 years) indicating they were in late childhood or adolescence in order to study the long term psychological sequelae of the disorder. They had their first operation at least 5 years before. Twenty of them were males (76.9%) and 6 were females (23.1%).

Only 2 patients had mild hypospadias (7.7%) while 18 patients (69.2%) suffered severe hypospadias and 6 patients (23.1%)

suffered virilisation. This does not mirror the prevalence of the disorders in general population⁶. Further, all patients underwent their 1st. corrective surgery at least 5 years ago, but they needed repeated surgical intervention mean 3.85 ± 1.74 st.d. (Range: 2-9). It may reflect that long term follow up is not naturally available except for more severe forms who actually still suffer.

Outcome of surgery was investigated from the surgeon' view point we found that 92.3% (n=24) improved. 30.8% (n=8) having incontinence as the only functional problem while 84.6% (n=22) having residual cosmetic problem. This is comparable to most research on the subject which found that penile appearance and scars are inevitable complications of almost all surgical procedures¹³.

Parents' view of the outcome of surgery was investigated in order to assess satisfaction after surgery. 92.3% found they were improved but still have cosmetic and functional problems. This is different from an earlier study which found only 38% who still felt deformed¹⁸ but comparable to another study which found 72% regarded their penis as abnormal¹⁹. Difference may be due to the choice of sample. In our study, most patients (n=18) f (69.2%) had severe hypospadias.

Both surgeon and parents' views were comparable ($p=0.03$), indicating that parents were as objective as the surgeons in perceiving their children's condition. This implies that assessment of parents' psychological adjustment was not towards an imagined problem.

In our study, Results showed that the surgical condition was upsetting in 38.5% (n=10). This is comparable to previous studies where adolescents operated on for

hypospadias often experience negative genital appraisal where they are less satisfied with the appearance of their genitalia (20-21); On the other hand, negative view of self was the least encountered in our sample although still a substantial number 23.1% (n=6). This is different from previous studies where adolescents operated on for hypospadias and the comparison group both had an equal, positive score for self concept²¹. Moreover, social avoidance was found in 38.5% (n=10) patients. This is comparable to previous studies where adolescents often felt less accepted by peers than comparison groups yet difference was not of statistical significance²¹. Another study showed that those children suffer more emotional problems and that this resulted from harassment⁵.

We found no research investigating the influence of ambiguous genitalia or genitoplasty outcome on parents' psychological adjustment specifically. However, many research addressed parents' adjustment to child chronic illness and child physical disability or deformity. The long duration, painfulness and severe side effects of treatment and the uncertainty of disease progression often result in tremendous stresses to parents²². In a study investigating grief reactions of mothers of adolescents and young adults with traumatic brain injury, time -since-injury, within the 3 years post injury period appeared to significantly influence guilt as an aspect of maternal grief. Guilt was shown to be especially intense during the earliest and latest time periods or when attempting to regain a personal life routine in the face of perceived care-taking obligations²³. In our study guilt was only correlated significantly to number of surgeries ($P=0.02$) and to age at time of the

study ($P=0.04$) which reflects time-since-diagnosis. This finding could have the same explanation. In the same study, mothers' perception of their children's neurobehavioral functioning was of direct relevance to their own grief reactions²³. Similar to our study where diagnosis with more impact of severe hypospadias and virilisation rather than mild hypospadias on total guilt, denial, depressive symptoms and anxiety ($P=0.003$; 0.02 ; 0.0008 ; 0.001 respectively). Residual functional problems and residual cosmetic defect were highly significantly associated with all aspects of grief.

Various factors were investigated for possible risk for psychological maladjustment in children.

Gender: there was a highly significant difference in negative view of self between males and females being higher in females ($p=0.04$). There are not many studies about the psychosocial and sexual outcome of genital surgery in females born with ambiguous genitalia. In one study, it was found that difficulties in clitoral sensuality was correlated with communication difficulties and avoidance²⁴ or due to problems about gender behavior as most women with ambiguous genitalia are masculine in behavior as suggested by earlier research²⁵⁻²⁶. In another study, a total of 14 girls, mean age 13.1 years, with congenital adrenal hyperplasia were assessed under anesthesia by a pediatric urologist, plastic/reconstructive surgeon and gynecologist. Of these patients 13 had previously undergone feminizing genitoplasty in early childhood at 4 different specialist centers in the United Kingdom. The outcome of clitoral surgery was unsatisfactory (clitoral atrophy or prominent glans) in 6 girls, including 3

whose genitoplasty had been performed by 3 different specialist pediatric urologists. Additional vaginal surgery was necessary for normal comfortable intercourse in 13 patients. Fibrosis and scarring were most evident in those who had undergone aggressive attempts at vaginal reconstruction in infancy. The psychological issues surrounding sexuality in these patients are inadequately researched and poorly understood²⁷. This is contradictory to earlier studies which found that overall, most girls and women with CAH identify as female and feminizing surgery in the neonatal period is the standard^{8,14}.

Age of the patients during the study was highly and positively correlated to a degree to which appearance is upsetting and to avoidance ($p=0.0001$ for both). This is comparable to other research which found there is a significant effect of age on the body image: the older the children, the lower their body image²⁰⁻²¹.

Clinical diagnosis: There was a significant difference between mild to moderate hypospadias patients, severe hypospadias patients and virilization with respect to negative view of self and avoidance in patients being more in the more severe forms. Also, number of surgeries was positively and significantly correlated to upset by penile appearance, and avoidance. This was explained by the last relation where outcome of surgery as regards residual functional problem was very highly significantly associated with negative view of self and residual cosmetic defect which was very highly significantly associated with degree of upset with penile appearance and avoidance. These results are in accordance with a previous research where proximal hypospadias patients have more

negative appraisals to the results of hypospadias and penile appearance^{4,20}. They were more inhibited in seeking girlfriends or unwilling to go to bathroomhouses or public lavatories where they often hide their genitals because of embarrassment⁴.

Psychological adjustment in children was then correlated to grief in parents. Negative view of self was positively correlated to total grief of parents and results were of statistical significance ($P=0.01$). There was a positive correlation to depressive symptoms in parents and results were of high statistical significance ($P=0.002$). Upset about appearance of genitalia was positively correlated to anxiety of parents and results were of statistical significance ($P=0.02$). This result was not replicated in any previous study as none of previous studies took into account the impact of parent's psychological state on the child's psychological state.

An important limitation of the study was small sample size. This was due to difficulty in finding patients operated upon for ambiguous genitalia who are still available for follow up after a minimum of 5 years postoperative. Another limitation is that the sample recruited may be the patient group with the worst outcome among all patients operated upon for ambiguous genitalia and thus psychological maladjustment was most prevalent.

Despite limitations of the study, we can conclude from the results that long term outcome studies should always consider psychological wellbeing of the child as well as his parents with implications on the practice guidelines for those patients. A multidisciplinary team involving mental health service professionals is of utmost importance for managing psychological

maladjustment in children and their parents with the final aim of a better quality of life which is now considered an important outcome measure.

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